Open Access Emergency Medicine

Dovepress
Taylor & Francis Group

Open Access Full Text Article

CASE REPORT

A Case of Cutaneous Leishmaniasis Presenting to the Emergency Department

Anne Gordon[1], Adrienne N Malik[2]

[1]The University of Kansas School of Medicine, Kansas, Kansas, USA; [2]The University of Kansas Medical Center Emergency Department, Kansas, Kansas, USA

Correspondence: Anne Gordon, The University of Kansas Medical Center, 3901 Rainbow Boulevard, Kansas, KS, 66160, USA, Email agordon5@kumc.edu

Abstract: In this case, we describe a case of cutaneous leishmaniasis, a protozoan disease not typically found in the United States, that presented to our emergency department (ED). The diagnosis was confirmed by the Center for Disease Control and Prevention (CDC) through a polymerase chain reaction (PCR) sample from the presenting skin lesion. The patient was a 43-year-old woman with history of a bite by an unknown organism while traveling by foot through Panama. She presented with a large, ulcerated lesion on her lower left shin. She was admitted on suspicion of leishmaniasis, and physicians of several specialties initiated a broad laboratory workup, collected wound cultures, prescribed antibiotics, and surgically repaired the lesion. The rapid recognition of leishmaniasis allowed for the patient to begin treatment before the definitive diagnosis returned, emphasizing the importance of knowledge of worldwide diseases and their presenting features for the ED physician.

Keywords: case report, rash, parasite, vector-borne disease

Introduction

Leishmaniasis is a vector-borne protozoan disease caused by the *Leishmania* parasites.[1] This disease is transmitted to humans via a bite of the female sand-fly Phlebotomus (Africa, Asia, Europe) or Lutzomyia (New Continent).[1] It is endemic in East Africa, Latin America, and Southeast Asia in forested areas and there are approximately 1.5–2 million new cases and 70,000 deaths each year.[2,3]

There are several forms of leishmaniasis including localized cutaneous, diffuse cutaneous, mucosal, and visceral leishmaniasis.[3] Visceral leishmaniasis is the most lethal and the least common as it is only transmitted by two species of Leishmania. Over 90% of visceral cases occur in North Africa, South Asia, and Brazil.[2,4] Cutaneous leishmaniasis is a severe disease but usually not fatal.[1] It presents as ulcerative lesions that often spontaneously heal in 6–18 months.[1] 10% of cutaneous leishmaniasis cases evolve into more severe, systemic forms of the disease (mucosal, mucocutaneous) which are much more deadly.[1]

The *Leishmania* parasite enters human cell as promastigotes, the first stage of a parasite and are then phagocytized by cutaneous macrophages.[1] Usually, immunocompetent hosts are able to kill the promastigotes, and the parasite never spreads to other organs via cellular lysis.[1] However, if the promastigotes survive, they become amastigotes, divide and cause cellular lysis.[1] Leishmaniasis progresses into different clinical presentations, courses, and outcomes as amastigotes propagate into other reticular-endothelial system cells.[1]

The course and gravity of the disease depends on both the patient's immune system and the infecting species. Generally, the wound begins at the site of the vector injection and creates papules within two weeks.[3] Later, lymph nodes become involved and eventually a granuloma can evolve from this lesion.[3] The granuloma can then ulcerate and delay healing which often becomes chronic.[3]

Although leishmaniasis is endemic in certain areas, the increased migration of people and ecological changes has made leishmaniasis a more worldwide disease.[1,2] The systematic disease progression makes rapid diagnosis and

Received: 15 October 2024
Accepted: 14 January 2025
Published: 7 February 2025

treatment imperative to patient recovery. It is very challenging to make a diagnosis of leishmaniasis in the emergency department (ED) in the US due to a lack of familiarity with the disease and ability to test the blood or lesion for Leishmania.[2] This case report will demonstrate a care strategy based off leishmaniasis as the leading differential diagnosis as confirmation of *Leishmania* presence can take a long time to return from the Center for Disease Control and Prevention (CDC).

Case Report

A 43-year-old Venezuelan female presented to the ED in Kansas City, Kansas in February of 2024 with a large, ulcerated wound on her lower left shin spanning the width of her ankle (Figure 1). She had a history of getting bitten by an unknown organism while traveling by foot through Panama from Venezuela to the US. Since arriving in the US two months prior, the patient reported the wound worsened in appearance. She was previously seen in the San Philippe Clinic where she took amoxicillin followed by doxycycline, both of which yielded no improvement.

In the ED, the patient reported burning, intolerable pain that impeded her ability to walk. On physical exam the wound was found to have well-demarcated, ulcerated appearing tissue underneath raised, crusted tissue or what was thought to possibly be areas of fungal infection (Figure 1). The following labs were obtained: complete blood count with differential, comprehensive metabolic panel, c-reactive protein (CRP), lactate, and blood cultures. Significant findings included an elevated erythrocyte sedimentation rate (49), normal CRP, and normal lactate. A left lower extremity x-ray was obtained showing soft-tissue edema and no bony erosion or periosteal reaction to suggest osteomyelitis or retained foreign body.

After consultation with infectious disease the patient was admitted for extensive workup and management. She was seen by infectious disease, dermatology, and plastic surgery and started on cefepime on admission. A punch biopsy was performed the following day and pathology showed a chronic wound with dense lymphoplasmacytic infiltrate and poorly formed non-necrotizing granulomas. Acid-fast bacteria stains, human immunodeficiency virus, galactomannan, *Histoplasma, Coccidiodes, Fungitell, Blastomyces, Bartonella henselae* and *quintana* IgM and IgG were all negative. The biopsy tested positive for toxoplasma IgG but negative for IgM.

Six days after the first biopsy, plastic surgery excised the lesion and placed a cadaveric skin allograft. The pathology of the excised ulcer was analyzed, and a large skin lesion was sent to the CDC for polymerase chain reaction (PCR) testing. The wound culture returned with *Serratia liquefaciens* and *Finegoldia magna* growth. The use of rare stains yielded a positive EBV result however this was believed to be nonspecific. Following this data, the patient was changed to levofloxacin and metronidazole and then did a final course of oral penicillin V and levofloxacin which was completed

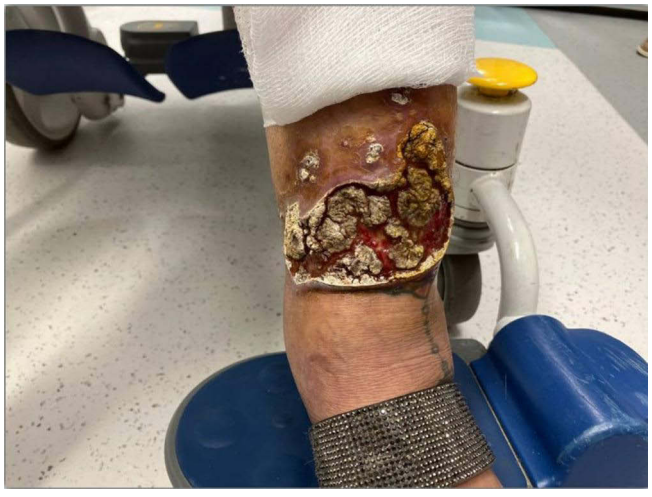

Figure 1 Leishmaniasis wound at presentation. This is a photograph of the patient's wound at the time of presentation to the emergency department (ED). There is an ulcerated lesion with crusted areas and adjacent soft tissue edema located on the anterior aspect of the left lower shin. There did not appear to be adjacent bony erosion or periosteal reaction to suggest osteomyelitis on same-day x-ray.

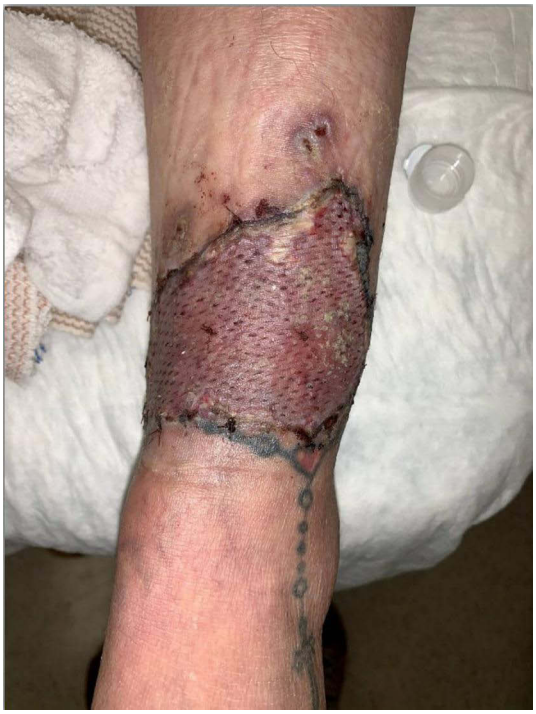

Figure 2 Leishmaniasis Lesion after Skin Autograft. The patient returned to the operating room 10 days after the cadaveric allograft was placed. Plastic surgery removed the allograft and placed an autograft. The patient followed up with plastic surgery 11 days after the autograft placement (image show).

one day after discharge. The patient returned to the operating room after ten days for allograft removal and autograft placement (Figure 2). She was discharged four days post-autograft placement.

After the patient was discharged, the final diagnosis from the CDC PCR of the large skin lesion sample was complex cutaneous leishmaniasis from *Leishmania panamensis*. At her follow-up visit, the patient was treated in accordance with the CDC guidelines for her case. She received a combination of oral Miltefosine (2.5 mg/kg/day with a maximum of 150 mg, in 3 divided doses for 28 days) and liposomal Amphotericin B (IV 3 mg/kg/d for Days #1-5, then another dose on day #10). After Amphotericin B was completed, the patient received a 28-day course of oral Ketoconazole (600 mg daily). Her treatment and dosing was in line with CDC published guidelines.[5] Miltefosine was approved in 2014 by the FDA and is indicated for three species, one of which includes *Leishmania panamensis*.[5] Ketoconazole and other "azoles" have shown variable results however ketoconazole showed modest efficiency against *Leishmania panamensis*.[5] The CDC lists the lipid formulations of amphotericin B as a potential treatment for CL but the evidence supporting its use for CL is limited to case reports and series.[5]

The species *Leishmania panamensis* can cause mucosal leishmaniasis and could disseminate from CL. Therefore, the patient was counseled per the CDC's advice, on the signs of mucosal leishmaniasis and instructions to seek medical attention should these arise. She was informed that the CDC does not believe, despite her surgical procedures, that a sterile cure for leishmaniasis was achieved. The patient was advised about the possibility of reactivation should she become immunocompromised or reinjure the lesion site.

Discussion

In this case, the patient presented to the ED with a severely ulcerated, painful skin lesion consistent with leishmaniasis. She was treated empirically with antibiotics and surgical resection before the diagnosis of leishmaniasis was made via PCR at the CDC. There is currently no rapid diagnostic tool widely available and the methods for diagnosing leishmaniasis vary. The historical diagnostic method is microscopic histopathological examination of biopsies of the lesion.[1,3] This method has a high specificity but the sensitivity ranges from 50% to 90% as it depends on the biopsied tissue itself.[1] PCR-based molecular diagnostic techniques have a 100% specificity, and the sensitivity is improved

compared to traditional parasitological diagnostic methods.[3] However, the widespread use of PCR diagnostic methods is limited by it high cost and requirement for technical expertise.

Our patient's diagnosis of *Leishmania panamensis* is unsurprising given her history of an insect bite while traveling on foot through jungle in Panama that caused the initial high index of suspicion for active disease at the time of her ED visit. This species of the *Leishmania* parasite is particularly prevalent in Panama and transmitted by multiple species of Lutzomyia sand flies that live across the country.[2] Additionally, this patient exhibited signs of superinfection as her lesion was painful and CL lesions usually are painless. The findings of Serratia liquefaciens and Finegoldia magna, necessitated the switch from cefepime to levofloxacin (Serratia liquefaciens coverage) and metronidazole (Finegoldia magna coverage).[6] The final course of oral penicillin V (Finegoldia magna coverage) and levofloxacin was implemented to ensure eradication of the superinfection.

The problem clinicians face when treating cutaneous leishmaniasis is that the drugs used to treat the disease are reserved for patients with a confirmed diagnosis of leishmaniasis. They are not given on clinical diagnosis due to their serious side effects including renal failure, hepatotoxicity, cardiotoxicity, and musculoskeletal pains.[3] Pentavalent antimony, given parenterally or intralesionally, is the first-line treatment for cutaneous leishmaniasis. However, according to the CDC, these medications are not approved by the Food and Drug Administration and are thus unavailable in the United States.[5] After this patient was definitively diagnosed with Leishmaniasis, she was treated with miltefosine, liposomal amphotericin B, and ketoconazole. Notably, studies have shown that treatment efficacy widely varies by infecting species. Miltefosine was shown to be 91% effective and ketoconazole was 76% effective in treating L. panamensis, the culprit species of our patient's infection.[3]

This case highlights the need to consider leishmaniasis when assessing victims of human trafficking or recent immigrants from endemic areas who may have been without care for some time prior to being seen in the ED. While this patient eventually received care, this case underscores the need for a need for further research into a more curative treatment and vaccine for leishmaniasis given its propensity to reactivate and spread systemically.[3] The current research on vaccines for *Leishmania* has remained futile. Currently, several types of vaccines against *Leishmania* are being studied including DNA adjuvant and recombinant antigen vaccines but no vaccine has been used clinically in humans yet.[7] As we become a more global community with immigration and frequent travel, it is ever more vital for physicians to recognize and know how to treat international illnesses.

Abbreviations

ED, emergency department; CDC, Center for Disease Control and Prevention; CRP, c-reactive protein; PCR, polymerase chain reaction.

Ethics Approval and Informed Consent

The patient was informed and consented to the use of the included pictures and medical information for publication. The University of Kansas Medical Center Institutional Review Board (IRB) considers case reports and case series exempt from needing IRB approval. Therefore, IRB approval was not required to publish this case.

Disclosure

The authors report no conflicts of interest in this work.

References

1. Piccioni A, Valletta F, Zanza C, et al. Rapid clinical management of leishmaniasis in emergency department: a case report with clinical review of recent literature. *Biology*. 2020;9(11). doi:10.3390/biology9110351
2. Dutari LC, Loaiza JR. American cutaneous leishmaniasis in panama: a historical review of entomological studies on anthropophilic Lutzomyia sand fly species. *Parasite Vectors*. 2014;7(1):218. doi:10.1186/1756-3305-7-218
3. Reithinger R, Dujardin JC, Louzir H, Pirmez C, Alexander B, Brooker S. Cutaneous leishmaniasis. *Lancet Infect Dis*. 2007;7(9):581–596. doi:10.1016/S1473-3099(07)70209-8
4. Pace D. Leishmaniasis. *J Infect*. 2014;69(Suppl 1):S10–S18. doi:10.1016/j.jinf.2014.07.016
5. Clinical Care of Leishmaniasis. Center for disease control and prevention. [Updated March 13, 2024]. Available from: https://www.cdc.gov/leishmaniasis/hcp/clinical-care/index.html. Accessed August 4, 2024.

6. Antibiotic Stewardship and Spectrum Guide. Infectious diseases management program at UCSF; July 2021. Available from: https://idmp.ucsf.edu/content/antibiotic-stewardship-and-spectrum-guideUpdated. Accessed November 27, 2024.
7. Kelleci K, Allahverdiyev A, Bağirova M, Ihlamur M, Abamor E. Particulate and non-particle adjuvants in Leishmaniasis vaccine designs: a review. *J Vector Borne Dis*. 2023;60(2):125–141. doi:10.4103/0972-9062.361179